Large Unilateral Noduloplaque Over the Cheek in a Young Man

Case History

A 30-year-old man presented with a large asymptomatic nodule over the right cheek that appeared 5 years back and enlarged gradually. Patient's primary concerns were suspicion of a malignancy and facial cosmetic disfigurement. History of prior local trauma or systemic medication was absent. Current history of photosensitivity, and food- or beverage-induced erythema/flushing was negative. Past medical, surgical, and family histories were noncontributory. On examination, the nodule was observed to be skin-colored, 5 × 6 cm in size, and localized over the right cheek inferior to the right lower eyelid. The firm and nontender nodule was immobile and fixed to the underlying structures, with a smooth overlying skin [Figure 1A]. Mild erythema, few acneiform papules, and atrophic scars (suggestive of healed acne) were seen over both the cheeks. There was no visible central punctum or pore. Dermoscopic examination of the plaque revealed few enlarged pilosebaceous openings but no telangiectasias. The lesion was excised *in toto*. Clinical reevaluation after 6 months revealed no recurrence, although the overlying acne scars seemed more conspicuous, possibly due to the release of the swelling-induced stretching force [Figure 1B]. The histopathological examination from the excised lesion revealed multiple circumscribed, well-developed, and separated sebaceous gland lobules with several normal and occasional improperly oriented vellus follicles in the mid dermis (hematoxylin and eosin, ×100) [Figure 2]. There was no cystic change and no infiltrative growth pattern, lymphovascular space invasion, cytologic atypia, or mitotic figures. Collagen bundles in adventitial dermis were thickened with an increased number of dilated thick-walled capillaries in upper-to-mid dermis (hematoxylin and eosin, ×400) [Figure 3]. Immunohistochemical staining with antibody to Ki-67 nuclear antigen was found to be negative.

Diagnosis

Giant solitary sebaceous gland hyperplasia.

Discussion

Facial sebaceous gland overactivity associated with inflammation is typically encountered in acne and rosacea. Less commonly, a solitary noninflammatory

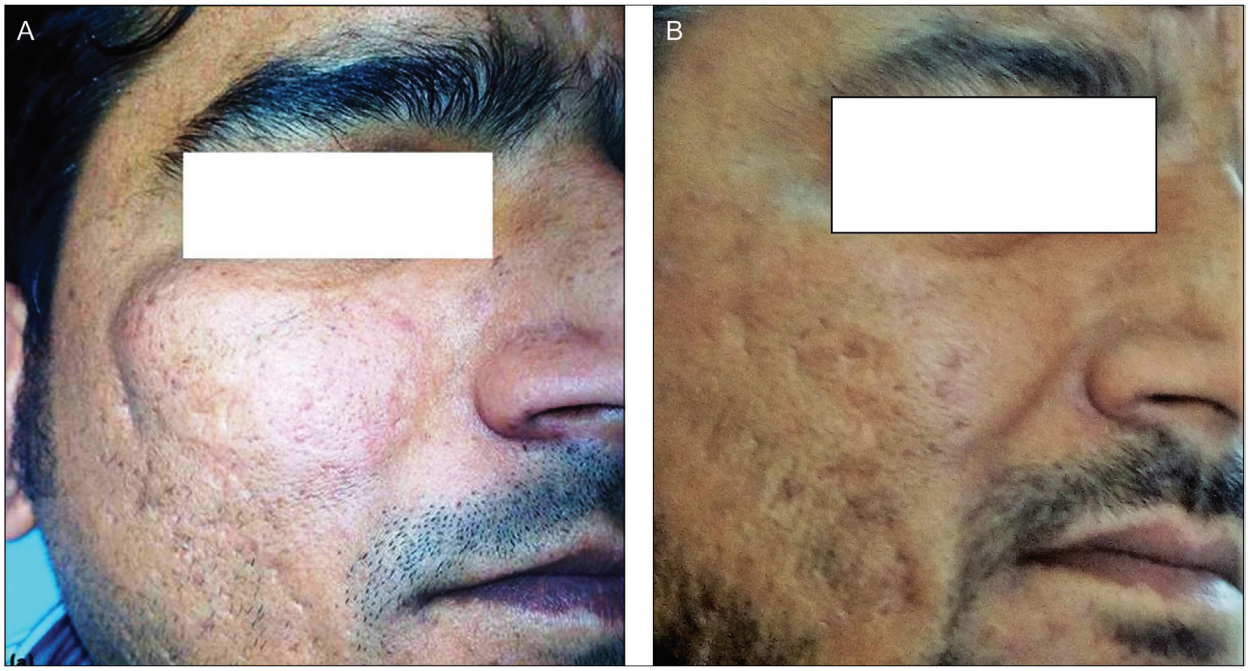

Figure 1: Clinical picture of the patient revealing a 5 × 6 cm sized, skin-colored, firm, non-tender nodule with smooth surface over the right cheek (A), and 6 months after successful complete excision (B)

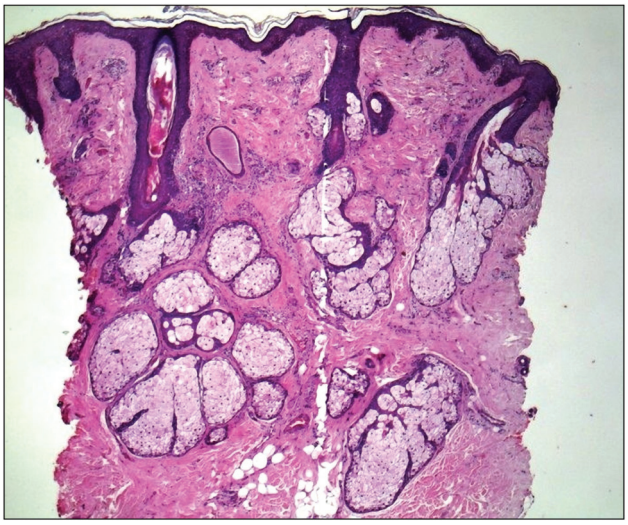

Figure 2: Histopathology revealing several normal and occasional improperly oriented vellus follicles in mid dermis with numerous well-developed sebaceous glands with absence of any epithelial or appendageal neoplasm (hematoxylin and eosin, ×100)

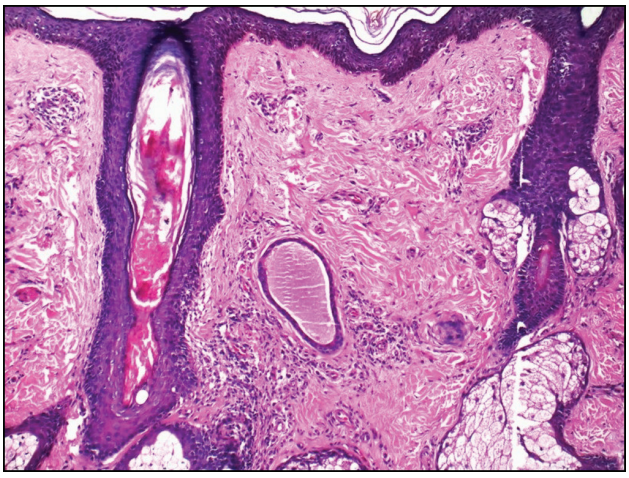

Figure 3: Histopathology at higher magnification revealing thickened collagen bundles in adventitial dermis with dilated thick-walled capillaries in upper-to-mid dermis (hematoxylin and eosin, ×400)

sebaceous gland hyperplasia may develop resulting in a painless facial nodule. Giant solitary sebaceous gland hyperplasia (GSSGH) is a rare nonneoplastic sebaceous gland lesion that presents as a large solitary intracutaneous nodule and may involve the frontal scalp, nose, cheeks, and vulva.[1-3] It presents as a skin-colored, firm, non-tender nodule with a smooth surface that may display surface umbilications. Histologically, a sharply demarcated hyperplasia of grouped mature sebaceous glands is characteristic and a conservative proliferating pattern confirms its benign hyperplastic character.[1,2] Clinical differential diagnoses include sebaceous neoplasms such as sebaceous adenoma, sebaceoma, and sebaceous carcinoma (extraocular). Other differentials include epidermoid cyst and folliculosebaceous cystic hamartoma (FSCH).

In contrast to sebaceous hyperplasia that is composed of only aggregates of mature sebaceous glands and lacks any germinative basaloid cells, sebaceous neoplasms consist of a variable combination of basaloid cells admixed with sebocytes and sebaceous ductlike structures located in the dermis with or without connection to the epidermis.

A sebaceous adenoma, in addition to sebaceous gland lobules that are also seen in sebaceous hyperplasia, consists of variably expanded basaloid cells (more than the normal two-cell layers seen in normal sebaceous glands and sebaceous hyperplasia). However, the majority of this benign tumor is still composed of sebaceous lobules, with the proportion of basaloid cells never exceeding that of sebocytes. A sebaceous adenoma may develop cystic degeneration in the center; such a tumor being typically encountered in Muir–Torre syndrome. A sebaceoma is a well-circumscribed lobulated tumor, composed majorly of basaloid cells and few sebaceous cells. A sebaceoma also frequently demonstrates areas of cyst formation and frequent mitoses, although cytologic atypia is absent.[4]

Malignant sebaceous gland carcinoma most commonly arises in the periocular area as a pink or yellowish nodule with only a few reported cases of extraocular site involvement. It has documented capability of regional and distant metastasis and high rates of recurrence after excision.[5] Histological features are suggestive of a malignant process, including presence of cytologic atypia, areas of necrosis, deep extension, and adnexal or lymphovascular invasion.

An epidermoid cyst, a flesh-colored to yellowish, adherent, firm, round nodule, typically with a central pore or punctum is very often considered in the differential diagnosis of such lesions.[1] Histology is characterized by unilocular or multilocular cystic structures occupying the dermis, containing lamellated and acellular keratin.

FSCH is a distinct cutaneous hamartoma that typically involves the nose, cheeks, forehead, and the scalp. Some consider FSCH to be a variant of sebaceous trichofolliculoma. Although an FSCH may mimic solitary sebaceous hyperplasia clinically, it is distinguishable by its distinctive histology. Unlike solitary sebaceous hyperplasia, FSCH consists of cystic structures that are lined with stratified squamous epithelium and show well-developed, dilated infundibular portions of hair follicles with infundibular keratinization, with mature sebaceous lobules radiating from these cystic structures. In addition, a prominent mesenchymal component in the form of densely laminated collagen bundles with vascular proliferation, and variable increases in muscle components around the cystic structures are typically present.[6]

Thus, the lesional morphology, its benign clinical course, and a typical histology characterized by well-demarcated hyperplasia of grouped mature sebaceous glands, a proliferating but noninfiltrative growth pattern, lack of cystic

structures, and absence of lymphovascular space invasion, cytologic atypia, and mitotic figures firmly established the diagnosis of this huge lesion as GSSGH. Complete surgical excision remains the mainstay of GSSGH therapy.

Five Learning Points

- Solitary sebaceous gland hyperplasia is a rare nonneoplastic sebaceous gland lesion that presents as a solitary intracutaneous nodule and may involve the frontal scalp, nose, cheeks, and vulva. It may attain gigantic proportions when it may be called a GSSGH.
- In contrast to sebaceous neoplasms, its close clinical differentials, a well-demarcated hyperplasia of grouped mature sebaceous glands and a conservative proliferating pattern on histology, make its differentiation possible.
- Neoplasms like sebaceous adenoma and sebaceoma are characterized by replacement of sebocytes with basaloid cells, presence of cystic degeneration, and presence of mitotic figures.
- Other differentials include epidermoid cyst and folliculosebaceous cystic hamartoma, with the latter displaying cystic structures lined with stratified squamous epithelium and presence of well-developed, dilated infundibular portions of hair follicles with infundibular keratinization.
- Complete surgical excision remains the mainstay of GSSGH therapy.

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Conflict of interest

None.

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